The benefits and enjoyment of a swimming intervention for youth with cerebral palsy

Pediatric Physical Therapy

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Abstract:	Purpose To investigate enjoyment and specific benefits of a swimming intervention for youth with cerebral palsy (CP). Methods Fourteen youth with CP (aged 7 to 17 years, GMFCS I to III) were randomly assigned to control and swimming intervention groups. Walking ability, swimming skills, fatigue and pain were assessed at baseline, after a 10-week swimming intervention (2/week, 40-50 minutes) or control period, after a 5-week follow-up and, for the intervention group, after a 20-week follow-up period. The level of enjoyment of each swim-session was assessed. Results Levels of enjoyment were high. Walking and swimming skills improved significantly more in the swimming than control group ($p = .043$; $p = .002$), whilst fatigue and pain did not increase. After 20 weeks gains in walking and swimming skills were retained ($p = .017$; $p = .016$). Conclusion We recommend a swimming program for youth with CP to complement a physical therapy program.

Abstract

Purpose

To investigate enjoyment and specific benefits of a swimming intervention for youth with cerebral palsy (CP).

Methods

Fourteen youth with CP (aged 7 to 17 years, GMFCS I to III) were randomly assigned to control and swimming intervention groups. Walking ability, swimming skills, fatigue and pain were assessed at baseline, after a 10-week swimming intervention (2/week, 40-50 minutes) or control period, after a 5-week follow-up and, for the intervention group, after a 20-week follow-up period. The level of enjoyment of each swim-session was assessed.

Results

Levels of enjoyment were high. Walking and swimming skills improved significantly more in the swimming than control group (p = .043; p = .002), whilst fatigue and pain did not increase. After 20 weeks gains in walking and swimming skills were retained (p = .017; p = .016).

Conclusion

We recommend a swimming program for youth with CP to complement a physical therapy program.

Introduction

Cerebral palsy (CP) is the most common motor disability in childhood and is associated with lifelong motor impairment. [1] Diverse impairments of body function and structure in addition to activity limitations and participation restrictions have been identified in youth and adults with CP; Seventy per cent of the European youth with CP are able to walk with or without aids, [2] but at lower walking speed than in typically developing youth. [3] Reduced walking speed can limit the ability of children and adolescents with Gross Motor Function Classification Scale (GMFCS) levels II and III to keep up with peers, especially outdoors and in the community. [4] Additionally, 45% of adults with CP reported a deterioration of walking skills, with an onset of deterioration between 15 and 34 years of age for 64% of these adults. [5] Secondary problems developing mainly in late childhood include fatigue and pain. [6] Pain has been reported to be present in 60% of 8 to 12 year-olds with CP [7] and in 74% of 13 to 17 year-olds with CP. [8] Thirty per cent of adults with CP report substantial perceptions of fatigue, and their reported physical fatigue is significantly higher than in the general population. [9] Adolescents and young adults with CP perceive the physical therapy program during childhood to induce fatigue, pain and physical distress. [10,11]

Physical activity has been found to contribute significantly to the prevention of chronic pain, fatigue and deterioration of locomotor skills in adults with CP. [11] Whilst sustaining a physically active lifestyle is essential (for youth with CP) to achieve and maintain functional capability, [12] youth with CP are considerably less active than their able-bodied peers. [13] Perceived barriers to engaging in physical activity reported by youth with CP include fatigue, pain during exercise, fear of increased risk of injury, beliefs that learning a motor skill is too time-consuming and the perception of physical activity and sports as not

being fun. [14] A lack of physical activity has been found to be associated with perceived physical fatigue and to contribute to the deterioration of locomotion in adults with CP. [5,9] Since an increase in pain and perceptions of fatigue is associated with a higher chance of inactivity among adults with CP with the ability to walk, [15] a vicious cycle of inactivity exists.

Motivation and enjoyment are known to be facilitators for engaging in physical activity and for adhering to physical activity. [10,16] A recent review of Riner and Sellhorst [12] recommended physical activity and exercise programs for youth with CP to be enjoyable, to be within the child's capabilities, and to include only activities with limited risk of falling or injury. Swimming is a community-based exercise that is believed to be fun, not to increase pain during exercise and not to increase the risk for injury in youth with CP, [17-19] but no randomized controlled trial has been published to support this so far. Swimming and other aquatic interventions have been reported to have a positive effect on gait velocity [18,20] and aquatic skills. [17-19] Kelly, et al. [21] reported that fatigue was not significantly increased after a 12-week community aquatic exercise program. Levels of pain associated with the aquatic intervention have not been reported in any of these studies. Moreover, none of the authors reported the perceived level of enjoyment of the participants regarding the intervention programs.

Therefore, the purpose of this study was to investigate the effect of a swimming intervention on pain, fatigue, walking ability, and aquatic and swimming skills, in youth with CP with the ability to walk, and the retention of possible gains. Furthermore, the enjoyment of the swimming program was evaluated.

Method

The study used a randomized controlled design with single blinding. Youth diagnosed with CP, aged 7 to 17 years, with GMFCS levels I to III were recruited through hospitals, special schools and private practices. Exclusion criteria were a botulinum toxin A injection or orthopedic surgery during the 6 months prior to the start of the study. Parents provided full informed consent. Ethical approval was obtained from the university hospital medical ethical committee (Trial number: S53366). Participants were randomly assigned to a control group (no scheduled swimming program) or an intervention (swimming) group. Randomization was blocked by age (< 12.5 y & ≥ 12.5 y) and by GMFCS level (I, II & III).

The 10-week swimming program in the community was offered without financial cost to the participants in the intervention group[i] and consisted of two sessions per week (range 40 – 50 min) in a 25 m by 13 m swimming pool (27.5 °C). The immediate objective of the program was to improve independence in the water and to learn or improve a swimming stroke. Participants in the intervention group were individually tutored and some activities were carried out in group with others present in the pool. A maximum of 4 participants were together in the pool at any one time. The main investigator instructed the youth assisted by physiotherapy students. Details of the swimming intervention program can be found in the appendix. Immediately after each swimming session the participants rated their perceived level of enjoyment of the swimming session on a 5-point Likert scale using smiley faces and labels ranging from ‘not at all’, ‘a little’, to ‘very much’. All participants

[i] For ethical reasons, the intervention program was also offered without financial cost to the participants in the control group (after the 5-week follow-up tests for this study had taken place).

(of intervention and control group) continued to receive their usual physical therapy programme throughout the study, which was reported to the main investigator and did not differ between groups (Table 1).

A 5-week follow-up period with no scheduled swimming program for either group followed. All participants were evaluated three times: before (T_1) and after (T_2) the intervention/control period and after the 5-week follow-up period (T_3). The swimming group was assessed once more, 20 weeks after the end of the swimming program. The three assessors had a BSc in Physiotherapy and Rehabilitation Sciences and were blinded as to group assignment. All assessors were trained in administration of the tests and followed a protocol for administration to ensure consistent instructions. All assessors each assessed an equal number of participants of each group to avoid bias.

The main investigator, aware of the group assignment, conducted the measurements in the water. Participants in the control group took part in all tests, including the pool-based measurements. Self-reported current pain intensity and the amount of hurt or pain in the past week were measured using the Faces Pain Scale - Revised (FPS-R) and the Visual Analogue Scale (VAS), respectively. Both tools are valid and reliable and the combination of scales is considered the most appropriate for use in clinical trials in children and adolescents. [22] The user-friendly 1-minute fast walk test (1-min WT) measured distance walked at maximum walking speed and is valid and reliable for use in children with CP. [23] Perceptions of fatigue were measured using the Dutch version of the self-report 'PedsQL™ multidimensional fatigue scale' (PedsQL Fatigue), [24] which is valid and reliable for use in children and adolescents. [25] The Water Orientation Test Alyn 2 (WOTA 2), a 27-item test based on the Halliwick concept [26], assessed the swimmer's level of

adjustment and function in the water. The scale is reliable and valid for use in youth with disabilities and consists of a mental adjustment (MA) subscale and a skills, balance control and movement (SBM) subscale. [27]

Data analyses were performed using SPSS v19 and Microsoft Excel 2010. The α-level was set at .05 and all tests were two-tailed. Demographics and characteristics at baseline were compared between the swimming and control group using Mann-Whitney *U* tests for the ordinal (GMFCS, Manual Ability Classification Scale (MACS), etc.) and continuous (age, anthropometrics, etc.) descriptive measures and Fisher's exact tests for the categorical descriptive measures (swimming experience (y/n), use of orthotic devices (y/n)). The baseline values of the outcome variables were compared between groups using a Mann-Whitney *U* test or an unpaired *t*-test. Changes over the 10-week (T_1 to T_2) and the 15-week period (T_1 to T_3) were compared between groups using Mann-Whitney *U* tests for the pain intensity scores, PedsQL Fatigue scores and the WOTA 2 scores, and unpaired *t*-tests for the 1-min WT scores. Changes over time (T_1 - T_2 - T_3) within each group were analyzed using a Friedman's two-way analysis of variance by ranks for pain intensity, the PedsQL Fatigue and the WOTA 2. Where significant results were found by the Friedman's two-way ANOVA, post-hoc tests corrected for multiple comparisons were used to test the differences between baseline (T_1) and post-test (T_2) and between baseline (T_1) and follow-up test (T_3). Changes over time within each group for the 1-min WT were analyzed using a repeated measures analysis of variance (simple contrasts, first). Bonferroni correction for multiple testing was applied. The differences between the baseline values and the 20-week follow-up scores of the swimming group were evaluated using Wilcoxon matched-pairs signed rank tests and paired *t*-tests.

Results

The participant flow diagram is shown in Figure 1. The number of possible candidates assessed for eligibility is unknown, as some therapists and institutions did not reveal the number of eligible participants due to privacy regulations. Of the 40 individuals who responded to the recruitment efforts, 15 were randomized and completed the baseline testing, and 14 (7 control and 7 intervention) completed the study and were included in the data analysis. One participant of the control group dropped out due to a persistent viral infection. The majority of participants was classified in GMFCS level II (n = 10) and the 7-12.5 year (n = 10) categories.

The two groups were not significantly different at baseline in demographics, characteristics and physical ability (Table I); however the participants in the control group were slightly older, heavier and taller than participants in the intervention group. Both groups were comparable at baseline for the outcome measurements.

Adherence and enjoyment

All participants of the intervention group completed 16 to 20 swimming sessions (median adherence 100%). All individuals but one rated their levels of enjoyment with a median maximum score (5), indicating that the swimming sessions were enjoyed 'very much'. One child reported a score of 3, indicating that the sessions were enjoyed 'a little bit'. No adverse events related to the study procedures were reported.

Walking ability

One adolescent of the control group was unable to perform the 1-min WT at post-test (T_2), due to a knee injury unrelated to the study. The walking distance at maximum walking

speed of the swimming group improved over time (T_1 - T_2 - T_3), but not to a level of significance (Table II). Over the 10-week swimming intervention (T_1 to T_2), the improvement in walking distance at maximum walking speed of the swimming group was significantly different from the change in the control group (Table III). No significant differences were observed between groups for their changes over the 15-week period (T_1 to T_3) that included a 5-week follow-up period. However, walking distance in the swimming group increased by 18.9 m compared to a 4.9 m increase in the control group. After the 20-week follow-up period, the 1-min WT scores of the swimming group were significantly higher than at baseline, with a mean improvement of 14.6 m (Table IV).

Pain

One participant's pain intensity data (swimming group) were removed from the analysis because of inability to comprehend the pain intensity scales, as judged by the assessor in consultation with the participant's parents and school teacher. Baseline values of both the VAS and the FPS-R scale were low (Table II). There was a high variability for the pain intensity scores within each group and between the two measurement scales. Changes over time were not significantly different between groups and no significant change over time within either group was reported (Table II). Between-subject variability for the change in pain intensity between the measurement occasions was high.

Perceptions of fatigue

Two participants' data of the PedsQL Fatigue (swimming group) were removed from the analysis because of inability to comprehend the questionnaire, as judged by the assessor in consultation with the participants' parents and schoolteachers. The PedsQL Fatigue scores

of the swimming group did not change significantly over time, while the PedsQL Fatigue scores of the control group did change significantly over time (Table II); post-hoc tests revealed a significant increase in fatigue between baseline (T_1) and follow-up (T_3) in the control group. The changes over the 10- and 15-week periods did not differ between the swimming and the control group (U = 16.0, Z = -0.245, p = .874; U = 9.0, Z = -1.390, p = .199, respectively). Although not reaching significance, after 20 weeks follow-up, the PedsQL Fatigue scores of the swimming group were higher than at baseline (representing a decrease in fatigue), with a median improvement of 4% (Table IV).

Swimming skills

The total score, the MA subscore and the SBM subscore of the WOTA 2 changed significantly over time in the swimming intervention group (Table II). The results of the post-hoc tests in this group revealed significant improvements from baseline (T_1) to post-intervention (T_2) and from baseline to the end of the 5-week follow-up period (T_3) for all scores. In contrast, the control group showed no significant change in the total score or the MA subscore over the test period. The SBM subscore of the control group changed significantly over time, and post-hoc tests revealed a significant improvement from baseline (T_1) to post-test (T_2) (16.7% absolute increase). It should be noted, however, that in the swimming group all WOTA 2 scores increased significantly more than in the control group for both the 10- and 15-week periods (Table III). After the 20-week follow-up period, the swimming group's total score and both subscores of the WOTA 2 remained significantly higher than the baseline values (Table IV).

Discussion

The aim of this study was to investigate the effect of a swimming intervention on pain intensity, perceptions of fatigue, walking ability and aquatic and swimming skills, in youth with CP with the ability to walk (with or without hand-held mobility devices), and the retention of possible gains. There is a paucity of studies in the literature investigating the influence of swimming programs on the various impairments and activity limitations encountered by youth with CP. Among barriers for youth with CP to engaging in physical activity is the perception that physical activity and sports are not fun. [14] Therefore, the perceived level of enjoyment of the swimming intervention was also assessed.

All youth in the swimming intervention group had a high adherence to the swimming program and reported high levels of enjoyment. Swimming skills improved after the 10-week program and walking ability showed a trend towards improvement, without adverse effects on pain intensity and fatigue. These gains in the swimming intervention group were retained 20 weeks after the end of the program.

Walking ability

The findings show that one of the indicators of walking ability, walking distance at maximum walking speed, increased in the swimming intervention group after the 10-week swimming program. The change in walking distance over the 10-week swimming program was significantly different from the change over this period in the control group. Twenty weeks after the completion of the swimming program, a significant increase from baseline was retained in the swimming intervention group. Maximum walking speed has not been reported in any other study on aquatics in youth with CP. Changes larger than 5.1m were

deemed clinically relevant [23]; such clinically relevant changes were found for three participants after the swimming intervention and for five participants 20 weeks after completing the swimming intervention. As walking with restrictions limits the ability of youth to keep up with peers in the community, [4] the improvement in walking speed can facilitate participation in the community.

Fatigue and pain

Self-reported feelings of fatigue did not increase in the swimming intervention group after the 10-week swimming intervention, which is in agreement with the findings of Kelly, et al. [21], who indicated no changes in fatigue after a 12-week aquatic exercise program in a sample of five 9 to 11 year-old children with CP. In the present study, fatigue increased significantly in the control group over the 15-week period (T_1 to T_3). Important to note are the high baseline levels of the sample in the present study (sample median of 89.9%), which indicates that feelings of fatigue occurred rarely in the month prior to baseline. These high scores imply that the youth of the present sample did not feel fatigued frequently. There was a high variability for the changes in pain intensity scores within each group and between the two measurement scales. The changes over time for self-reported pain intensity did not differ significantly between groups. These findings are in contrast to regular physical therapy programs that have been reported to cause pain and physical distress. [10] Pain and fatigue are perceived by youth with CP to increase due to exercise and consequently are barriers to engaging in physical activity.[14] Additionally, increases in pain and fatigue have been reported to be associated with a higher chance of inactivity in adults with CP. [15] Therefore it is pertinent that the engagement in the physical activity program in the present study did not increase levels of pain or fatigue. It is

interesting to note that the relatively cold water in the community swimming pool apparently did not affect pain or fatigue.

Swimming skills

Swimming skills improved significantly more in the swimming intervention group over the 10-week swimming program than in the control group, and the changes were retained with significance after a 20-week follow-up period. During this follow-up period, four of the seven participants of the swimming group swam during their school time, or recreationally with family. Swimming skills improved by more than the minimal detectable change (14.2%) [27] in six of the seven participants in the swimming intervention group. The improvement in swimming skills after the swimming intervention supports previous research. [17-19] However, in these studies no control group had performed the aquatic tests nor was a follow-up period longer than 3 weeks included. [17-19] The strong retention found in the present study implies that the swimming skills were learned and consolidated during the 10-week intervention. This contrasts with the perception that learning a motor skill is too time-consuming for youth with CP [14] and is important with regard to the retention of swimming skills that enables the participants to engage in a greater variety of physical activities performed in the water.

Enjoyment and adherence

All participants of the swimming intervention group but one reported enjoying the swimming sessions ‘very much’ (maximum score of 5). By participating in the swimming program the youth experienced that sport and exercise programs, in this case swimming, can be fun. Therefore, a swimming program such as the one offered in this study has the

ability to eliminate one of the barriers to engaging in physical activity. [14] Since enjoyment and motivation are important facilitators for engaging in physical activity, adhering to therapy, and sustaining a physically active lifestyle, [10,12,16] the findings of the present study are important. The suggested positive relationship between enjoyment and adherence is supported by the high attendance rates (median 100%) found in the present study. Furthermore, none of the participants dropped out during the intervention, implying that the motivation to complete the swimming program was high.

Limitations

Only 14 participants completed the study and some participants were not capable of completing the pain intensity scales and the PedsQL Fatigue questionnaire, which negatively affected the power of the statistical analysis and increased the possibility of type II errors. Due to practical reasons, the assessor of the WOTA 2 was not blinded to group assignment. However, the WOTA 2 test was evaluated according to the objective criteria as explained in the manual [27]. Another limitation of the study is the lack of reliability and validity studies of the use of the pain and fatigue measures in youth with CP; however the measures are psychometrically sound for use in children. [22,25] The data of those showing difficulties with completing the pain measures and the PedsQL were excluded. The motor performance measures (WOTA 2 and 1-min WT) have been tested for validity and reliability in youth with CP, [23,27] however the cognitive ability of the children was not specified in these studies. Due to the nature of the measure, we did not experience any difficulties with the instructions regarding the motor tasks. Another limitation of the study is the use of convenience sampling, which might have contributed to the high enjoyment levels in this sample.

Nevertheless, the present study showed that the commonly perceived barriers to physical activity participation [14] were non-existent in the case of the swimming program, since in addition to high enjoyment during the program, levels of fatigue or pain did not increase due to the swimming program, and swimming skills improved and were retained. These factors are important with regard to the sustainment of a physically active lifestyle. [12] Additionally, while participating in this physically active swimming program, motor proficiency on land and in the water improved. A swimming intervention is therefore a recommended physical activity for ambulatory youth with CP to combat the vicious cycle of inactivity and improve mobility both in and out of the water.

Conclusion

Specialists, physical therapists and parents should become aware of the benefits of a swimming program for ambulant youth with CP, as it could complement a rehabilitation program. Physical activity and sport programs should be promoted to youth with CP with trial and introductory sessions provided in community-based settings in collaboration with physical therapists, as a lack of information sustains the perception that physical activity and sport programs are not enjoyable and increase pain and fatigue. Finally, it is highly recommended to assess the perceived level of enjoyment in any intervention program, since it closely relates to adherence.

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Figure legend

Figure 1. Diagram illustrating the flow of participants and group assignment, including the number for each level of the Gross Motor Function Classification System (GMFCS), within each age category. One child of the control group dropped out at T_2 due to a persistent viral infection.

Dear reviewers and editor,

Thank you very much again for your time and your helpful comments. We believe we have addressed all the reviewers' suggestions, by adding information to enhance clarity, as well as by rewording relevant sentences. Our reaction to your comments is given below.

Reviewer #1: Page 3- line 7 "adult walkers" should be changed to "adults with the ability to walk" as stated in line 47 on this page. This was in your original manuscript but I must have overlooked it.

> *In the sentence:* "Since an increase in pain and perceptions of fatigue is associated with a higher chance of inactivity among adult walkers with CP, [14] a vicious cycle of inactivity exists.", "Adult walkers*" has been changed to* "adults with the ability to walk*". The sentence on page 3 (line 11) now reads*: "Since an increase in pain and perceptions of fatigue is associated with a higher chance of inactivity among adults with CP with the ability to walk, [15] a vicious cycle of inactivity exists."

Page 4- lines 47-52. You state that all participants continue to receive their usual care. I would still like to see a statement about whether the subjects were receiving OT/PT or community programs during this time. In your responses to the reviewers comments you state that "levels of participation in these activities varied between participants but were not different between groups" I suggest you include this statement in the manuscript and refer readers to Table 1 which includes the number of PT minutes/week for each group.

> *This information has been added to the sentence* "All participants (of intervention and control group) continued to receive their usual care throughout the study, which was reported to the main investigator and documented.*", so the sentence now reads (page 4, line 55 - continued on page 5):* "All participants (of intervention and control group) continued to receive their usual physical therapy programme throughout the study, which was reported to the main investigator and did not differ between groups (Table 1)."

Page 5- lines 12 and on- The other reviewer expressed a concern about reliability and I agree that this is a valid concern that should be addressed in the manuscript. Although you have no intra or inter reliability data available, in your response to reviewer's comments, you give an explanation of the steps you took to ensure high reliability of the data. You state you have not described all of these steps in the manuscript due to the word limit but I still think it is important to include at least a statement or two about this.

> *We took several steps to ensure high reliability of the data, a statement about this has been added in the manuscript to the paragraph:*
>
> "A 5-week follow-up period with no scheduled swimming program for either group followed. All participants were evaluated three times: before (T_1) and after (T_2) the intervention/control period and after the 5-week follow-up period (T_3). The swimming group was assessed once more, 20 weeks after the end of the swimming program. The three assessors had a BSc in Physiotherapy and Rehabilitation Sciences and were blinded as to group assignment."
>
> *The paragraph on page 5 (lines 13–30) now reads as follows:*
>
> "A 5-week follow-up period with no scheduled swimming program for either group followed. All participants were evaluated three times: before (T_1) and after (T_2) the intervention/control period and after the 5-week follow-up period (T_3). The swimming group was assessed once more, 20 weeks after the end of the swimming program. The three assessors had a BSc in Physiotherapy and Rehabilitation Sciences and were blinded as to group assignment. All assessors were trained in

administration of the tests and followed a protocol for administration to ensure consistent instructions. All assessors each assessed an equal number of participants of each group to avoid bias."

Page 11- line 42- Did you gather information on whether any of the subjects continued with a swimming program on their own? If so, I think this should be included.

For ethical reasons, the participants were not prohibited from swimming recreationally throughout the study period, however they were requested to report information on activities performed during the study period. During the 20-week follow-up period (of the swimming group), three participants of this group swam during their school time, less than, or equal to, once every 14 days, and one child swam with his father regularly. These 4 participants improved (n = 1), deteriorated (n = 2) or did not change (n = 1) over this follow-up period. The remaining three participants that did not engage in swimming during the follow-up period, showed small improvements during this 20-week follow-up period.
A sentence to include the additional information has been added to the paragraph:
"Swimming skills improved significantly more in the swimming intervention group over the 10-week swimming program than in the control group, and the changes were retained with significance after a 20-week follow-up period"

So the paragraph now reads (page 12, lines 13–24):

"Swimming skills improved significantly more in the swimming intervention group over the 10-week swimming program than in the control group, and the changes were retained with significance after a 20-week follow-up period. During this follow-up period, four of the seven participants of the swimming group swam during swam during their school time, or recreationally with family."

<u>Reviewer #2:</u> Abstract, Conclusion: ...CP to complement a (not 'the') physical therapy program.

As suggested, the sentences "We recommend a swimming program for youth with CP to complement the physical therapy program." *in the abstract, and* "Specialists, physical therapists and parents should become aware of the benefits of a swimming program for ambulant youth with CP, as it could complement part of the intensive rehabilitation program." *in the conclusion, have been adjusted, and now read as follows:*
Abstract (page 1, line 43): "We recommend a swimming program for youth with CP to complement a physical therapy program. **"**
Conclusion (page 14, line 30): "Specialists, physical therapists and parents should become aware of the benefits of a swimming program for ambulant youth with CP, as it could complement a rehabilitation program."

Introduction, pg 2: I still find the introduction awkward. The first paragraph is a listing of research. I suggest an opening sentence or two, to frame the research.

An introductory sentence has been added to the first paragraph of the introduction:

"Seventy per cent of the European youth with CP are able to walk with or without aids [1], but a lower walking speed than in typically developing youth is apparent. [2] Reduced walking speed can limit the ability of children and adolescents with Gross Motor Function Classification Scale (GMFCS) levels II and III to keep up with peers, especially outdoors and in the community. [3] Additionally, 45% of adults with CP reported a deterioration of walking skills, with an onset of deterioration between 15 and 34 years of age for 64% of these adults. [4] Secondary problems developing mainly in late childhood include fatigue and pain. [5] Pain has been reported to be present in 60% of 8 to 12 year-olds with CP [6] and in 74% of 13 to 17 year-olds with CP. [7] Thirty per cent of adults with CP report substantial perceptions of fatigue, and their reported physical fatigue is significantly higher than in the general population. [8] Adolescents and young adults with CP perceive the physical therapy program during childhood to induce fatigue, pain and physical distress. [9, 10] **"**

So the paragraph now reads as follows (page 2, lines 7–42):

"Cerebral palsy (CP) is the most common motor disability in childhood and is associated with lifelong motor impairment. [1] Diverse impairments of body function and structure in addition to activity limitations and participation restrictions have been identified in youth and adults with CP; Seventy per cent of the European youth with CP are able to walk with or without aids, [2] but at lower walking speed than in typically developing youth. [3] Reduced walking speed can limit the ability of children and adolescents with Gross Motor Function Classification Scale (GMFCS) levels II and III to keep up with peers, especially outdoors and in the community. [4] Additionally, 45% of adults with CP reported a deterioration of walking skills, with an onset of deterioration between 15 and 34 years of age for 64% of these adults. [5] Secondary problems developing mainly in late childhood include fatigue and pain. [6] Pain has been reported to be present in 60% of 8 to 12 year-olds with CP [7] and in 74% of 13 to 17 year-olds with CP. [8] Thirty per cent of adults with CP report substantial perceptions of fatigue, and their reported physical fatigue is significantly higher than in the general population. [9] Adolescents and young adults with CP perceive the physical therapy program during childhood to induce fatigue, pain and physical distress. [10,11]"

Pg 5, line16: suggest assignment (not 'allocation')

*'*Allocation*' has been replaced by '*assignment*' throughout the manuscript (page 5 on lines 25 and 33; page 13, line 36), as well as in Figure 1 and the figure legend (page 19).*

Pg 7, line 4: What was the reason the participant dropped out? Be specific, 'unrelated to the study' raises a question of judgement.

*The sentence "*One participant of the control group dropped out for reasons unrelated to the study.*" has been rewritten and now reads (page 7, line 19; and in the figure legend on page 19):* "One participant of the control group dropped out due to a persistent viral infection."

line 7: I suggest including (n=?) for both the GMFCS level II and for 7-12.5 years old categories

The number of participants has been included in the sentence: "The majority of participants was classified in GMFCS level II and the 7-12.5 year categories." *The sentence now reads on page 7 (line 22):* "The majority of participants was classified in GMFCS level II (*n* = 10) and the 7-12.5 year (*n* = 10) categories."

line 42: swimming group improved but not to a level of significance during this study (not 'non-significantly over time). Just need one parenthesis with a semicolon between T3 and Table II?

*We have corrected the sentence: "*The walking distance at maximum walking speed of the swimming group improved non-significantly over time (T1 - T2 - T3) (Table II).*" It now reads (on page 7, line 59 – continued on page 8, line 4): "*The walking distance at maximum walking speed of the swimming group improved over time (T_1 - T_2 - T_3), but not to a level of significance (Table II)."

Pg 8, line 13: How did you determine that the participant was unable to comprehend the scales? How did you determine the others did?
Line 36: Same question as above, how was this determined?

One participant did not respond when an example question was asked. In consultation with the parents and the school teacher it was decided that the child would not be able to respond reliably to the questions about pain (the VAS and the FPS-R) and fatigue (PedsQL Fatigue). If instructions were given, the child did respond with a correct motor action (e.g. in the case of the swimming skills test). One other participant had difficulties comprehending the questions of the fatigue questionnaire, and in consultation with the parents and school teacher it was decided that the child would not be able to respond correctly to these questions, however, for the VAS and FPS-R scale it was decided that the child would be able to give a reliable response, as a simpler question was asked.

The paragraphs on pain and perceptions of fatigue from the previous manuscript:

"Pain

One participant's pain intensity data (swimming group) were removed from the analysis because of inability to comprehend the pain intensity scales. Baseline values of both the VAS and the FPS-R scale were low (Table II). There was a high variability for the pain intensity scores within each group and between the two measurement scales. Changes over time were not significantly different between groups and no significant change over time within either group was reported (Table II). Between-subject variability for the change in pain intensity between the measurement occasions was high.

Perceptions of fatigue

Two participants' data of the PedsQL Fatigue (swimming group) were removed from the analysis because of inability to comprehend the questionnaire. The PedsQL Fatigue scores of the swimming group did not change significantly over time, while the PedsQL Fatigue scores of the control group did change significantly over time (Table II); post-hoc tests revealed a significant increase in fatigue between baseline (T_1) and follow-up (T_3) in the control group. The changes over the 10- and 15-week periods did not differ between the swimming and the control group (U = 16.0, Z = -0.245, p = .874; U = 9.0, Z = -1.390, p = .199, respectively). Although not reaching significance, after 20 weeks follow-up, the PedsQL Fatigue scores of the swimming group were higher than at baseline (representing a decrease in fatigue), with a median improvement of 4% (Table IV)."

The paragraphs on pain and perceptions of fatigue on page 8 (line 27 and further) now read:

"Pain

One participant's pain intensity data (swimming group) were removed from the analysis because of inability to comprehend the pain intensity scales, as judged by the assessor in consultation with the participant's parents and school teacher. Baseline values of both the VAS and the FPS-R scale were low (Table II). There was a high variability for the pain intensity scores within each group and between the two measurement scales. Changes over time were not significantly different between groups and no significant change over time within either group was reported (Table II). Between-subject variability for the change in pain intensity between the measurement occasions was high."

"Perceptions of fatigue

Two participants' data of the PedsQL Fatigue (swimming group) were removed from the analysis because of inability to comprehend the questionnaire, as judged by the assessor in consultation with the participants' parents and schoolteachers. The PedsQL Fatigue scores of the swimming group did not change significantly over time, while the PedsQL Fatigue scores of the control group did change significantly over time (Table II); post-hoc tests revealed a significant increase in fatigue between baseline (T_1) and follow-up (T_3) in the control group. The changes over the 10- and 15-week periods did not differ between the swimming and the control group (U = 16.0, Z = -0.245, p = .874; U = 9.0, Z = -1.390, p = .199, respectively). Although not reaching significance, after 20 weeks follow-up, the PedsQL Fatigue scores of the swimming group were higher than at baseline (representing a decrease in fatigue), with a median improvement of 4% (Table IV)."

Pg 9, line 50: Among barriers for youth with CP versus Barriers for youth with CP

'Among*' has been added to the start of the sentence* "Barriers for youth with CP to engaging in physical activity include the perception of physical activity and sports as not being fun. [13]*", and the*

sentence on page 10 (line 19) now reads: "Among barriers for youth with CP to engaging in physical activity is the perception that physical activity and sports are not fun. [14]"

Line 55: enjoyment assessed but not reported? Where is this in your results?

The levels of enjoyment are reported in the first section of the results on page 7 (lines 40–50): "All individuals but one rated their levels of enjoyment with a median maximum score (5), indicating that the swimming sessions were enjoyed 'very much'. One child reported a score of 3, indicating that the sessions were enjoyed 'a little bit'. No adverse events related to the study procedures were reported*."*

A subheading ("Adherence and enjoyment") has been added on page 7 (line 37) to make these results stand out more clearly.

Discussion- I think this would benefit from some subheadings?

The headings '*W*alking ability', 'Fatigue and pain', 'Swimming skills', 'Enjoyment and adherence', *and* 'Limitations' *have been added above the paragraphs on the relevant subjects in the discussion on pages 10 to 13.*

Pg 11, line 28: It is interesting to note… (not Interesting to note…) & Line 31 the community swimming pool… (not this community…)

The sentence "Interesting to note is that the relatively cold water in this community swimming pool apparently did not affect pain or fatigue.*" has been adjusted and now reads (page 11, line 60 – continued on page 12):* "It is interesting to note that the relatively cold water in the community swimming pool apparently did not affect pain or fatigue."

Pg 12, line 38: Add Limitations as a heading

The heading 'Limitations' *has been added on page 13 (line 22).*

Pg 13, line 37: …of inactivity and improve mobility both in and out of the water.

The suggested addition has been included in the sentence: "A swimming intervention is therefore a recommended physical activity for ambulatory youth with CP to combat the vicious cycle of inactivity*."*

The sentence now reads (page 14, line 19): "A swimming intervention is therefore a recommended physical activity for ambulatory youth with CP to combat the vicious cycle of inactivity and improve mobility both in and out of the water."

Line 46: Could this not complement all programs? Or just intensive? Suggest: …as it could complement a rehabilitation program (remove part of the intensive)

As suggested, we rewrote the sentence, "*S*pecialists, physical therapists and parents should become aware of the benefits of a swimming program for ambulant youth with CP, as it could complement part of the intensive rehabilitation program.", so *it now reads (page 14, line 30):* "Specialists, physical therapists and parents should become aware of the benefits of a swimming program for ambulant youth with CP, as it could complement a rehabilitation program."

One additional change has been made, to improve the quality of the sentence on page 5 (line 35): "Participants in the control group participated in all tests, including the pool-based measurements." *has been changed to* "Participants in the control group took part in all tests, including the pool-based measurements."

The benefits and enjoyment of a swimming intervention for youth with cerebral palsy: an RCT study.

Authors: Marlies Declerck, PT, PhD; Martine Verheul, PhD; Daniel Daly, PhD; Ross Sanders, PhD

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Conflict of Interest statement: The authors declare no conflict of interest.

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Running head: Swimming benefits youth with CP

At the time the data was collected for this research article, Marlies Declerck was a PhD student at the Institute for Sport, Physical Education and Health Sciences at the University of Edinburgh, Edinburgh, United Kingdom.

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Dear Dr. VanSant,

Thank you for accepting the manuscript pending minor revisions. We believe we have addressed the suggestions by adding information and rewording sentences to enhance clarity. Please find a detailed description of the comments and changes in the attachment.

Sincerely,

Marlies Declerck, PT, Ph.D.

Sport, Physical Education and Health Sciences,

University of Edinburgh

declerckmhp@gmail.com

Table I. Descriptive information at baseline for the participants of two groups of youth with cerebral palsy (*N* = 14).

	Swimming group (*n* = 7)	Control group (*n* = 7)	Statistics	*p*-value
Demographics				
Gender (male/female)	5/2	3/4	Fisher's test	.592
Age at enrolment (y:mo)	8:7 (3:4)	11:8 (3:5)	U = 19.0	.535
Age categories				
7 – 12.5 y	5	5	U = 24.5	1.00
12.5 – 17 y	2	2		
Mobility level				
GMFCS I	1	2	U = 24.0	1.00
GMFCS II	6	4		
GMFCS III	0	1		
Manual ability level				
MACS I	1	1	U = 24.5	1.00
MACS II	4	4		
MACS III	2	2		
CP subtype				
Unilateral spasticity	2	3		
Bilateral spasticity	4	2		
Dyskinetic	0	2		
Non – classifiable	1	0		
Gestational age (weeks)	33 (12)	39 (3)	U = 14.5	.220
Anthropometrics				
Height (m)	1.29 (0.3)	1.51 (0.3)	U = 18.5	.478
Weight (kg)	26 (13.0)	35 (16.5)	U = 15.0	.245
Related Medical History				
ASD	2	1		
Speech difficulties	0	2		
Visual impairment	5	4		
Hearing difficulties	0	0		
Seizure disorder	2	2		
Education				
Special needs education	7	5	Fisher's test	.462
Mainstream education	0	2		
Swimming experience	6	7	Fisher's test	1.00
Orthotic devices	7	6	Fisher's test	1.00
Physical therapy (min/week)	90 (60)	100 (45)	U = 24.0	.992

Note. Values are medians (inter-quartile range) for the continuous variables and are frequencies for ordinal and categorical variables. GMFCS: Gross Motor Function Classification System; MACS: Manual Ability Classification System; Cerebral palsy (CP) subtypes according to the Surveillance of CP in Europe; ASD: Autism Spectrum Disorder.

Table II. The results of the 1-minute fast walk test (1-min WT), the Visual Analogue Scale (VAS), the Faces Pain Scale Revised (FPS-R), the PedsQL™ multidimensional fatigue scale (PedsQL Fatigue) and the Water Orientation Tests Alyn 2 (WOTA 2) (total score, mental adjustment score (MA) and skills, balance control and movement score (SBM)) at baseline (T_1), post-test (T_2) and after 5 weeks of follow-up (T_3) for the swimming group (Exp) and the control group (Ctrl).

Outcome measurement		*n*	Baseline (T_1)	Post-test (T_2)	5-week follow-up (T_3)	Statistical analysis[b]	*p*-value
1-min WT (m)	Exp	7	69.1 (14.1)	80.7 (17.4)	88.0 (15.6)	$F(2,12) = 3.788$, MSE = 167.11	.053
	Ctrl	6	78.1 (20.2)	70.5 (32.1)	83.0 (29.7)	$F(2, 10) = 3.348$, MSE = 119.56	.077
VAS (0-100 mm)	Exp	6	6.5 (25.8)	12.0 (51.8)	13.5 (21.0)	$\chi^2(2) = 0.333$	.898
	Ctrl	7	18.0 (44)	28.0 (32)	6.0 (28.5)	$\chi^2(2) = 1.778$	.451
FPS-R (0-10)	Exp	6	1 (2)	0 (0)	0 (1.5)	$\chi^2(2) = 1.077$	.741
	Ctrl	7	0 (3)	2 (10)	0 (3)	$\chi^2(2) = 2.364$	.389
PedsQL Fatigue (%)[a]	Exp	5	76.4 (20.8)	73.6 (12.5)	83.3 (22.2)	$\chi^2(2) = 0.316$	.907
	Ctrl	7	94.4 (16.0)	91.7 (3.5)	88.9 (27.1)	$\chi^2(2) = 6.077$*	.042
WOTA total (%)	Exp	7	40.0 (5.6)	74.7 (16.9)	74.7 (18.3)	$\chi^2(2) = 13.000$**	<.001
	Ctrl	7	52.0 (23.5)	65.4 (31.9)	60.5 (36.3)	$\chi^2(2) = 4.571$	.112
WOTA MA (%)	Exp	7	60.6 (6.6)	87.2 (3.1)	90.9 (9.6)	$\chi^2(2) = 13.040$**	.001
	Ctrl	7	66.7 (25.1)	63.6 (35.2)	59.0 (43.8)	$\chi^2(2) = 0.240$	.932
WOTA SBM (%)	Exp	7	21.4 (7.1)	64.3 (33.3)	61.9 (33.3)	$\chi^2(2) = 11.385$**	.001
	Ctrl	7	35.7 (25.0)	66.7 (40.5)	61.9 (46.4)	$\chi^2(2) = 7.684$*	.019

Note. Values are medians (inter-quartile range) for all scales except for the 1-min WT (means and SD are presented). [a] An increase in PedsQL Fatigue represents a decrease in fatigue. [b] Changes over time were analyzed using a repeated measures ANOVA for the 1-min WT and a Friedman two-way ANOVA for the other scales. * $p < .05$; ** $p < .01$.

Table III. Comparison of the changes between the swimming and control group over the 10-week & the 15-week period for the 1-minute fast walk test (1-min WT) and the Water Orientation Test Alyn 2 (WOTA 2).

Outcome measurement	Swimming group	Control group	Statistical analysis[c]		*p*-value
1-min WT (m)	Mean (SD)		*t* [confidence interval]		
Baseline – post-test[a]	11.6 (18.2)	-8.3 (11.8)	2.289 [0.77 – 39.04]*		.043
Baseline – follow-up[b]	18.9 (23.1)	4.9 (17.5)	1.280 [-9.83 – 37.83]		.225
WOTA 2 total score (%)	Median (interquartile range)		*U*	*z*	
Baseline – post-test[a]	33.3 (8.7)	6.2 (9.3)	2.0**	-2.878	.002
Baseline – follow-up[b]	34.6 (9.3)	3.7 (9.1)	2.5**	-2.814	.003
WOTA 2 MA (%)	Median (interquartile range)		*U*	*z*	
Baseline – post-test[a]	24.2 (4.6)	0.0 (9.2)	0.0**	-3.148	.001
Baseline – follow-up[b]	30.3 (3.3)	-2.6 (13.3)	1.0**	-3.006	.001
WOTA 2 SBM (%)	Median (interquartile range)		*U*	*z*	
Baseline – post-test[a]	35.7 (17.9)	16.7 (21.4)	8.5*	-2.056	.042
Baseline – follow-up[b]	38.1 (15.5)	4.8 (20.2)	6.0*	-2.380	.016

[a]The values represent the absolute changes between the baseline score and the post-test score. [b]The values represent the absolute changes between the baseline score and the score after 5-weeks of follow-up. [c]Differences between groups were analyzed using unpaired *t*-tests for the 1-min WT and using Mann-Whitney *U* tests for the WOTA 2. TOT = total score; MA = mental adjustment subscore; SBM = skills, balance control and movement subscore. * $p < .05$; ** $p < .01$.

Table IV. The results of the 1-minute fast walk test (1-min WT), the Visual Analogue Scale (VAS), the Faces Pain Scale Revised (FPS-R), the PedsQL™ multidimensional fatigue scale (PedsQL Fatigue) and the Water Orientation Tests Alyn 2 (WOTA 2) (total score, mental adjustment score (MA) and skills, balance control and movement score (SBM)) at baseline and 20 weeks after the end of the program for the swimming group.

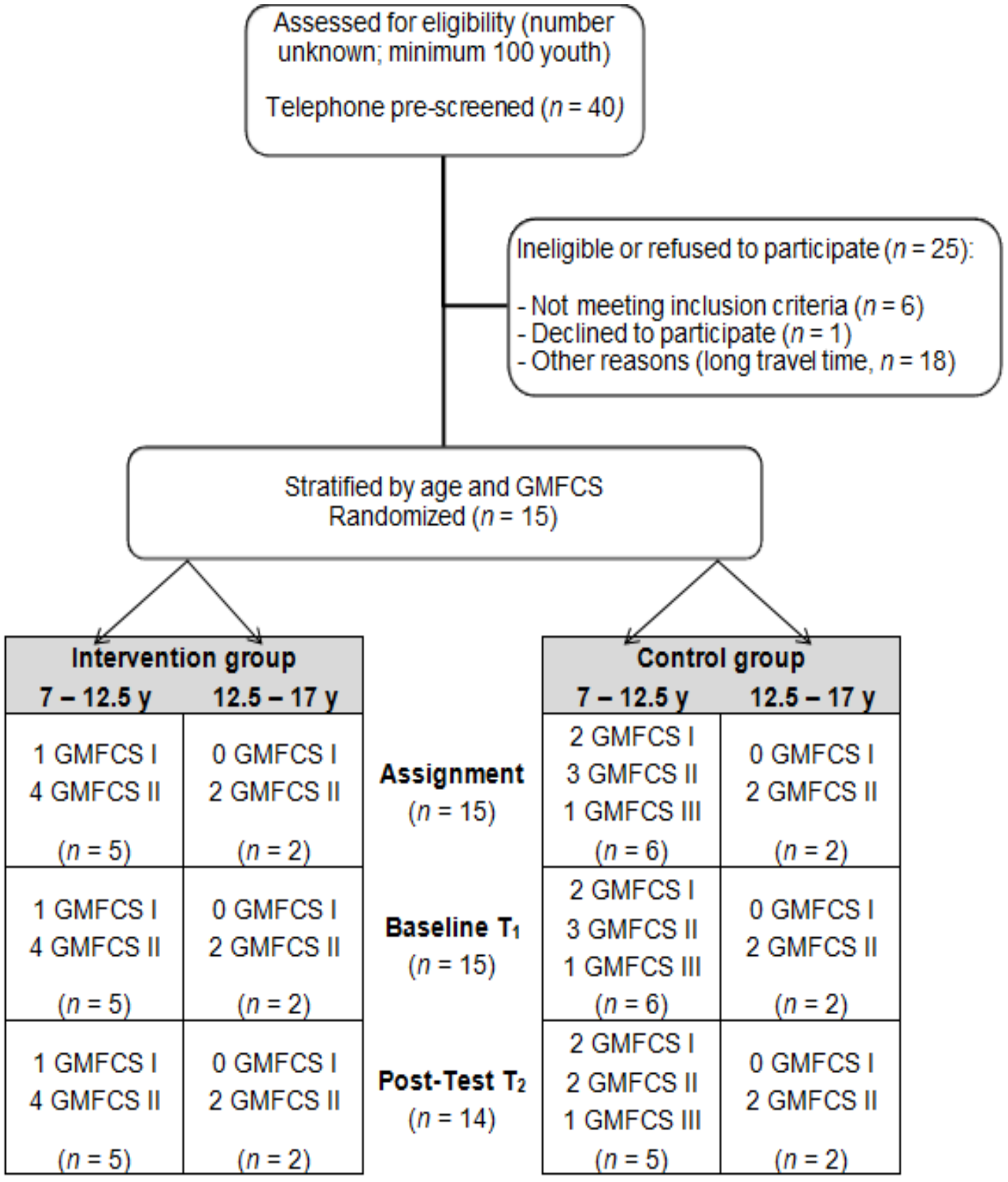
Outcome measurement	*n*	Baseline	20-week follow-up	Statistical analysis[b]	*p*-value
1-min WT (m)	7	69.1 (14.1)	83.7 (7.7)	$t(6) = 3.251$* CI [3.6 – 25.54]	.017
VAS (0-100 mm)	6	6.5 (25.8)	16.0 (41.8)	$Z = -1.095$	.375
FPS-R (0-10)	6	1 (2)	1 (3.5)	$Z = -0.813$	.750
PedsQL Fatigue (%)[a]	5	76.4 (20.8)	90.3 (8.3)	$Z = -2.023$	.064
WOTA total (%)	7	40.0 (5.6)	80.3 (24.9)	$Z = -2.366$*	.016
WOTA MA (%)	7	60.6 (6.6)	87.9 (12.1)	$Z = -2.371$*	.016
WOTA SBM (%)	7	21.4 (7.1)	69.1 (41.7)	$Z = -2.366$*	.016

Note. Values are medians (interquartile range) for all scales except for the 1-min WT (means and SD are presented). [a] An increase in PedsQL Fatigue represents a decrease in fatigue. [b] The differences between the baseline values and the 20-week follow-up scores of the swimming group were evaluated using a paired t-test for the 1-min WT and Wilcoxon matched-pairs signed rank tests for the other scales. * $p < .05$.

Appendix intervention details

Click here to download Supplemental Data File (.doc, .tif, pdf, etc.): Appendix Final 13 2.doc

Figure


Assessed for eligibility (number unknown; minimum 100 youth)
Telephone pre-screened (n = 40)
Ineligible or refused to participate (n = 25):
- Not meeting inclusion criteria (n = 6)
- Declined to participate (n = 1)
- Other reasons (long travel time, n = 18)
Stratified by age and GMFCS
Randomized (n = 15)
Intervention group
7 – 12.5 y
12.5 – 17 y
Control group
7 – 12.5 y
12.5 – 17 y
1 GMFCS I
4 GMFCS II
(n = 5)
0 GMFCS I
2 GMFCS II
(n = 2)
Assignment
(n = 15)
2 GMFCS I
3 GMFCS II
1 GMFCS III
(n = 6)
0 GMFCS I
2 GMFCS II
(n = 2)
1 GMFCS I
4 GMFCS II
(n = 5)
0 GMFCS I
2 GMFCS II
(n = 2)
Baseline T1
(n = 15)
2 GMFCS I
3 GMFCS II
1 GMFCS III
(n = 6)
0 GMFCS I
2 GMFCS II
(n = 2)
1 GMFCS I
4 GMFCS II
(n = 5)
0 GMFCS I
2 GMFCS II
(n = 2)
Post-Test T2
(n = 14)
2 GMFCS I
2 GMFCS II
1 GMFCS III
(n = 5)
0 GMFCS I
2 GMFCS II
(n = 2)